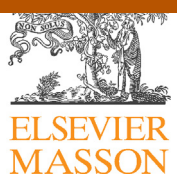
Orthopaedics & Traumatology: Surgery & Research (2013) 99S, S251—S254



Available online at

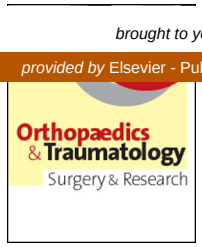
SciVerse ScienceDirect

www.sciencedirect.com

Elsevier Masson France

EM|consulte

www.em-consulte.com/en



WORKSHOPS OF THE SOO (2012, NANTES). ORIGINAL ARTICLE

Giant cell tumors of the tendon sheaths in the hand: Review of 96 patients with an average follow-up of 12 years

R. Lancigu[a,*], F. Rabarin[b], J. Jeudy[b], Y. Saint Cast[b], B. Cesari[b], P.A. Fouque[b], G. Raimbeau[b]

[a] *Département de chirurgie osseuse, CHU d'Angers, 4, rue Larrey, 49000 Angers, France*
[b] *Centre de la Main — Angers Assistance Main, 47, rue de la Foucaudiere, 49800 Trelaze, France*

KEYWORDS
Giant cell tumor;
Tendon sheath;
Recurrence;
Hand tumor

Summary
Introduction: Giant cell tumors (GCT) of the hand are relatively common and have a good prognosis, but the risk of recurrence is high. The goal of this study was to evaluate the long-term clinical results of a consecutive series of patients and to determine the risk factors for recurrence.
Material and methods: This was a retrospective study of 96 patients (57 women, 39 men) operated between February 1982 and October 2005 for GCT of the tendon sheaths in the hand. The average age at the time of the procedure was 47.7 ± 14.5 years (range 13—75). All the patients were reviewed by an independent surgeon. The following were recorded: clinical result (QuickDASH, satisfaction), recurrence, histological appearance of tumor, location of tumor, excision margins and extension into the neighboring anatomical structures (tendon, joint). The tumor was located in the index finger in 29 cases, middle finger in 23 cases, thumb in 21 cases, ring finger in 11 cases, little finger in 11 cases, hypothenar area in two cases and thenar area in one case. In all cases, the lesion was isolated. The swelling was palmar in 27 cases, dorsal in 20 cases and medial or lateral in 59 cases. The most common joint location was the DIP joint (35% of cases). The swollen area was sensitive in 12 cases. The time from the appearance of the tumor to physician consultation ranged between 1 month and 7 years. Before the surgery, standard X-rays were taken in all patients; ultrasonography was also performed in eight patients and MRI in one patient. The tumor had an average diameter of 15.8 ± 2.6 mm (range 5—30). Histological analysis revealed a multilobed lesion with multinucleated giant cells, with or without encapsulation.
Results: The average follow-up at the time of review was 12.1 ± 3.8 years (range 5—29). There were eight recurrences in seven patients (8.3%). The average time to recurrence was 2.75 ± 2 years (range 1—6.5). In every case of recurrence, there had been intra-articular tumor development and/or tendon destruction ($P < 0.01$). There was one functional complication: one DIP joint fusion secondary to one of the recurrences. The average QuickDASH was 2.3/100 (range 0—31).

* Corresponding author.
E-mail address: rlancigu@hotmail.fr (R. Lancigu).

1877-0568/$ - see front matter
http://dx.doi.org/10.1016/j.otsr.2013.03.008

Conclusion: Giant cell tumors of the synovial sheaths in the hand are benign lesions where recurrence is the primary risk. The recurrence typically occurred within 36 months of the excision. Intra-articular tumor development, marginal resection and tendon involvement seem to contribute to recurrence. There was no correlation found between the histological type of tumor (encapsulated or not) and recurrence.
Level of proof: IV.

Introduction

Giant cell tumors (GCTs) of the tendon sheaths in the hand are benign tumors that can invade the soft tissues of the hand and also the joints. These are one of the most common tumors of the hand, after synovial cysts [1]. GCTs have also been called myeloplax tumors, fibrous xanthoma, myeloxanthoma [2], and pigmented villonodularsynovitis [3]. The latter mostly involves large joints (knee, hip). All these named tumors have the same histological criterion: the presence of multinucleated giant cells. Although giant cell tumors (GCT) are considered benign and have a good prognosis, they have a high risk of recurrence [4—7]. The goal of this study was to evaluate long-term clinical results and determine risk factors for recurrence.

Material and methods

This was a retrospective study with 96 patients (57 women, 39 men) presenting a GCT of the tendon sheaths in the hand and operated between February 1982 and October 2005. The average age at the time of the procedure was 47 ± 15 years (range 13—75).

All the patients were evaluated again by an independent surgeon. A clinical questionnaire was completed over the telephone, as were the QuickDASH and a satisfaction questionnaire.

Statistical testing was performed with a Fisher's Exact test.

Before the surgery, standard A/P and lateral X-rays directly over the tumor were performed systematically. Ultrasonography was also performed in eight patients and magnetic resonance image (MRI) in one patient.

The surgical procedure was performed on an outpatient basis using regional anesthesia. A brachial tourniquet was used systematically and the procedure was carried out with surgical loupes.

The lesion was found in the index in 29 cases, middle finger in 23, thumb in 21, ring finger in 11, little finger in 11, hypothenar area in two and thenar area in one (Fig. 1). The most common GCT joint location was the DIP joint (35% of cases). The swelling was palmar only in 28% of cases, dorsal in 21% and medial or lateral in 51% of cases.

In 13.5%, an injury to the area had preceded the appearance of the lesion.

The time from the tumor appearance to physician consultation ranged between 1 month and 7 years.

The swollen area was sensitive in 12.5% of cases.

No other lesion was found on X-rays in 80 cases. Cortical deformation was visible in five cases and subchondral cysts in 11 cases (Fig. 2).

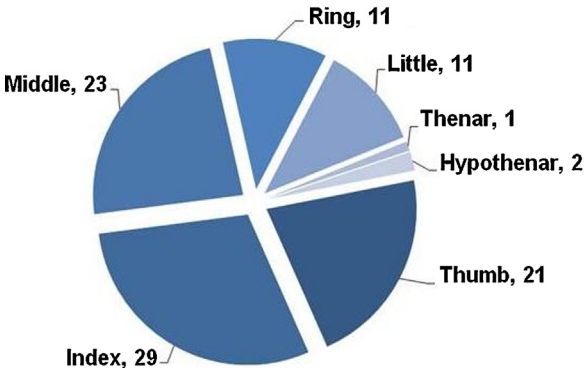


Figure 1 Distribution of tumors in the hand.

The ultrasonography assessment showed a hypoechogenic image that was not hypervascularized; the MRI showed a tissue with little gadolinium uptake.

The average size of the lesion was 15.8 ± 2.6 mm (range 5—30) determined by histological analysis. The lesion was encapsulated in 96% of cases. The tumor was multinodular in 62.5% of cases (Fig. 3) and was nodular in 47.5% of cases. Multinucleated giant cells were found in 100% of cases. Other cells such as histiocytes and siderophages were also found (Table 1).

Results

The average follow-up was 12.1 ± 3.8 years (range 5—29) (Table 2).

There were eight recurrences in seven patients (one patient had two recurrences despite the surgical

Figure 2 Subchondral cysts.

Figure 3 Multinodular lesion.

Table 1 Description of lesion.

Previous injury	13.5%
Tumor sensitivity	12.5%
Lesions visible on X-rays (*n*)	16
Average size (mm)	15.8
Most common joint localization	PIP
Capsule	96%
Multinodular lesion	62.5%

treatment). The average time to recurrence was 2.75 years (range 1—6.5).

There were two main, significant factors for the risk of recurrence:

- intra-articular lesion ($P = 0.002$);
- tendon erosion ($P = 0.01$).

Three other factors increased the risk, but not significantly:

- circumferential lesion ($P = 0.15$);
- multinodular lesions ($P = 0.12$);
- marginal excision ($P = 0.26$).

There were 12 patients with sequelae: six cases of stiffness with one patient requiring distal interphalangeal joint fusion and six cases of neurological sequelae such as paresthesia or hypoesthesia. The QuickDASH was 2.3/100 (range 0—31).

Upon being asked, 80% of patients indicated they were very satisfied, 19% were satisfied and 1% were partially satisfied with the outcome.

Table 2 Results.

Number of patients	96
Average age (years)	47
Average follow-up (years)	12
Recurrences (Number + %)	8 (8.3%)
Delay to recurrence (years)	2.75
Sequelae (Number + %)	12 (12.5%)
DASH at follow-up (/100)	2.3

Discussion

GCTs of the tendon sheaths of the hand are one of the more common soft tissue tumors in the hand after synovial cysts [1,8]. Its multiple names have been a hindrance to its definition. It was first described in 1852 by Chassaignac [9] who called it, ''malignant tumor of the tendon sheaths''. Then several other names were given to this tumor, such as ''myeloma of the tendon sheaths'' or ''myeloxanthoma'' [2]. The name ''giant cell tumor of the tendon sheath'' was introduced by Targett [10] and the name ''pigmented nodular synovitis'' by Jaffe et al. [3].

We found that GCTs mostly involve the first three fingers, with the index being most often involved, which is consistent with published data [4,8].

The patients had an average age of 47.7 years when the tumor was discovered. This was also consistent with published data reporting that this tumor mostly affects middle-aged adults [11,12] However, we had a 13 year-old patient in our series who presented with GCT but no recurrence. GCT is rarer in children [13—15].

Before surgery, a standard X-ray directly over the lesion should be performed [16]. Other than providing an opportunity to eliminate differential diagnoses, this will help to discover other lesions such as cortical erosion and subchondral cysts [7,17]. Ultrasonography usually detects a hypoechogenic lesion [18,19], although hyperechogenic have also been described [18,20,21]. MRI shows an low-signal lesion, mostly in the tissue [17,21].

Our 96 patient series revealed that GCTs are benign tumors that can recur. There was an 8.3% recurrence rate. This rate was low in comparison to certain publications that described a 30% rate [4—7]. Williams [22] found a 12.6% recurrence rate in a series of 213 patients and an average follow-up of 51 months. Al Qattan [23] found an 11.6% recurrence rate in a series of 43 patients with an average follow-up of 48 months. Looi et al. [11] described a 7% recurrence rate in his series with a follow-up of up to 60 months. In comparison, our study had a lengthy average follow-up and most of the recurrences appeared within 3 years. However, there was one recurrence at 78 months.

We found two parameters that were significantly related to recurrence — extension of the lesion into the intra-articular space ($P = 0.002$) (Fig. 4) and tendon erosion or invasion ($P = 0.01$). Williams [22] had also noted that these two factors were indicative of a high risk of recurrence (10%). We found other factors that were more common in recurring GCT in our series, although they were not significant predictors of recurrence: circumferential lesion, multinodular lesion, marginal excision. Others studies have shown an increased risk of recurrence for non-encapsulated tumors [4,23]. In our series, we found no evidence of cellular factors in recurrence, in contrast to other studies that showed the existence of elevated mitosis [24] and hypercellularity [1,25].

One of our patients had two recurrences, 3 years apart each time, despite complete excision and wide margins. Also, during the second recurrence a second tumor location was found (thumb interphalangeal joint and first phalanx of ring finger). No other specific factors, other than intra-articular localization and tendon erosion were found. At the

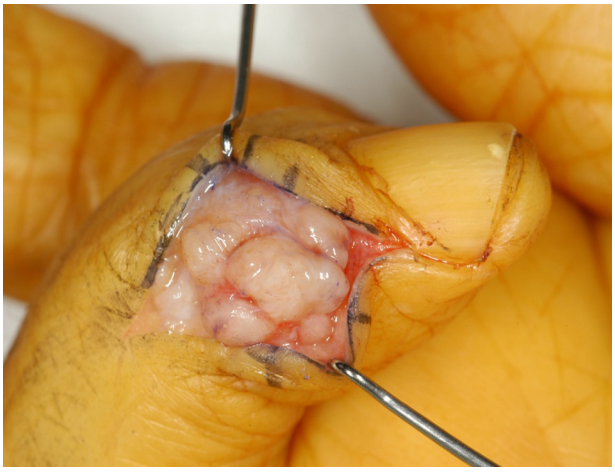


Figure 4 Intra-articular lesion.

last follow-up of this patient 16 years later, there had been no other recurrences. This double recurrence had not been described in published studies.

None of our patients required additional treatment. Although complete excision seems to be the best way to reduce the recurrence rate [1,11,26], some have suggested that postoperative radiation therapy could be effective in preventing recurrence [3,8] when risk factors are present or excision is incomplete. For example, the recurrence rate was reduced to 4% in the Kotwal study.

Although this is a benign tumor, it can lead to sequelae. We found a 12.5% rate of sequelae, which is consistent with other reports [27]. Six of our patients presented with dyesthesia at the final follow-up and six others had joint stiffness, which required PIP fusion in one patient.

Conclusion

GCTs of the tendon sheaths in the hand are a common tumor. They have a risk recurrence (8.3% in our series) that cannot be ignored. As a consequence, the patient must be informed of the possibility of this risk, even in cases of complete excision, and especially if the tumor extends into the joint or invades the tendons.

Disclosure of interest

The authors declare that they have no conflicts of interest concerning this article.

References

[1] Monaghan H, Salter DM, Al-Nafussi A. Giant cell tumor of tendon sheath (localised nodular tenosynovitis): clinicopathological features of 71 cases. J Clin Path 2001;54(5):404—7.
[2] Dor L. Relations des tumeurs à myeloplaxeset des xanthomes. Congres Chir 1898;12:553.
[3] Jaffe HL, Lichtenstein L, Sutro CJ. Pigmented villonodularsynovitis, bursitis and tenosynovitis. Arch Pathol 1941;31:371.
[4] Darwish FM, Haddad WH. Giant-cell tumor of tendon sheath: experience with 52 cases. Singapore Med J 2008;49(11):879—82.
[5] Moore JR, Weiland AJ, Curtis RM. Localized nodular tenosynovitis: experience with 115 cases. J Hand Surg [Am] 1984;9:412—7.
[6] Rao AS, Vigorita VJ. Pigmented villonodularsynovitis (giant cell tumor of the tendon sheath of the synovial membrane). A review of eighty-one cases. J Bone Joint Surg Am 1984;66:76—94.
[7] Reilly KE, Stern PJ, Dale JA. Recurrent giant cell tumors of the tendon sheath. J Hand Surg [Am] 1999;24:1298—302.
[8] Kotwal PP, Gupta V, Malhotra R. Giant-cell tumour of tendon sheath. Is radiotherapy indicated to prevent recurrence after surgery? J Bone Joint Surg Br 2000;82:571—3.
[9] Chassaignac M. Cancer de la gaine des tendons. Gazette Hop Civils Mil 1852;47:185—6.
[10] Targett JH. Giant cell tumors of the integument. Trans Pathol Soc Lond 1897;48:230—5.
[11] Looi KP, Low CK, Yap YM. Pigmented villonodularsynovitis of the hand in the Asian population. Hand Surg 1999;4:81—5.
[12] Ozalp T, Yercan H, Kurt C, Ozdemir O, Coşkunol E. Giant cell tumors of the tendon sheath involving the hand or the wrist: analysis of 141 patients. Acta Orthop Traumatol Turc 2004;38:120—4.
[13] Dartoy C, Fenol B, Leroy JP, Dubrana F, Le Nen D, Jehannin B. Tumeur à cellules géantes de la gaine du tendon du long fléchisseur du pouce chez une enfant de 7 ans. Ann Chir Main Memb Super 1994;13:198—201.
[14] Hulse N, Watson SJ. Giant cell tumour of tendon sheath in a child: a case report. Hand Surg 2005;10(1):97—100.
[15] Ueno T, Ansai S, Omi T, Kawana S. A child with giant cell tumor of tendon sheath. Dermatol Online J 2011;17(5):9.
[16] Uriburu IJ, Levy VD. Intraosseous growth of giant cell tumors of the tendon sheath (localized nodular tenosynovitis) of the digits: report of 15 cases. J Hand Surg [Am] 1998;23:732—6.
[17] Karasick D, Karasick P. Giant cell tumor of the tendon sheath: spectrum of radiological findings. Skeletal Radiol 1992;21:219—24.
[18] Fornage BD, Rifkin MD. Ultrasonic examination of the hand and foot. Radiol Clin North Am 1988;26:109—29.
[19] Martinoli C, Bianchi S, Derchi LE. Tendon and nerve sonography. Radiol Clin North Am 1999;37:691—711.
[20] Middleton WD, Patel V, Teefey SA, Boyer MI. Giant-cell tumors of the tendon sheath: analysis of sonographic findings. AJR Am J Roentgenol 2004;183(2):337—9.
[21] Peh WC, Truong NP, Totty WG, Gilula LA. Pictorial review: magnetic resonance imaging of benign soft tissue masses of the hand and wrist. Clin Radiol 1995;50:519.
[22] Williams J. Recurrence of giant cell tumors in the hand: a prospective study. J Hand Surg [Am] 2010;35:451.
[23] Al Qattan MM. Giant-cell tumours of tendon sheath: classification and recurrence rate. J Hand Surg [Br] 2001;26(1):72—5.
[24] Savage RC, Mustafa EB. Giant cell tumor of tendon sheath (localized nodular tenosynovitis). Ann Plast Surg 1984;13:205—10.
[25] Milack GP. Giant cell tumor of tendon sheath origin. A case report and literature review. J Am Podiatry Assoc 1983;73:643—5.
[26] Rodrigues C, Desai S, Chinoy R. Giant cell tumor of the tendon sheath: a retrospective study of 28 cases. J Surg Oncol 1998;68:100—3.
[27] Messoudi A, Fnini S, Labsaili N, Ghrib S, Rafai M, Largab A. Les tumeurs a cellules géantes des gaines synoviales de la main: a propos de 32 cas. Chir Main 2007;26:165—9.